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ORIGINAL ARTICLE

Predicting sequential bilateral cochlear implantation performance in postlingually deafened adults; A retrospective cohort study

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Objective: To identify which preoperative patient characteristics influence sequential bilateral cochlear implantation performance and to create a statistical model that predicts benefit.

Design: Multicentre retrospective cohort study.

Setting: All patients were operated in four academic teaching hospitals in Perth, Australia, and followed up by audiologists of the Ear Science Institute Australia.

Participants: A total of 92 postlingually deafened adult patients who had undergone sequential cochlear implantations between 19 June 1990 and 14 March 2016 were included. Patients were excluded if the 12-month follow-up consonant-nucleus-consonant (CNC) phoneme score was missing.

Main outcome measure: The effect of 18 preoperative factors on the CNC phoneme score in quiet (at 65 dB SPL) with the second cochlear implant (CI2) one year after implantation.

Results: Two factors were positively correlated to speech understanding with CI2: Wearing a hearing aid (HA) before receiving CI2 ($r = 0.46$, $P = 0.00$) and the maximum CNC phoneme score with the first CI (CI1) ($r = 0.21$, $P = 0.05$). Two factors were negatively correlated: the length of hearing loss before CI2 in the second implanted ear ($r = -0.25$, $P = 0.02$) and preoperative pure tone average (PTA) (0.5, 1, 2 kHz) before CI2 in the second implanted ear ($r = -0.27$, $P = 0.01$). The following model could be created: predicted CNC phoneme score with CI2 (%) = $16 + (44 * \text{HA use before CI2 (yes)}) - (0.22 * \text{length of hearing loss before CI2 (years)}) + (0.23 * \text{CNC phoneme score with CI1 (\%)})$. Because the effect of HA use before implantation played such a major role, we also created a model after exclusion of the HA factor: Predicted CNC phoneme score with CI2 (%) = $82 - (0.17 * \text{length of hearing loss before CI2 (years)}) - (0.27 * \text{PTA in second implanted ear before CI2 (0.5, 1, 2 kHz)}) + (0.20 * \text{CNC phoneme score with CI1 (\%)})$.

Conclusion: Advanced age or a long interval between implantations does not necessarily lead to poor CI2 results. Patients who are successful HA users before CI2, who have a low PTA before CI2, a high CNC phoneme score with CI1 and a limited length of hearing loss before CI2, are likely to be successful CI2 recipients.

1 | INTRODUCTION

Bilateral cochlear implantation offers advantages over unilateral cochlear implantation in patients with bilateral profound hearing loss. Bilateral implantation helps to restore sound localisation and improves hearing in noise and quality of life.^{1–8} Cochlear implant (CI) teams need to decide which patients are likely to benefit from a second CI and which patients are not. In general, they will consider a patient's age, duration of deafness, cause of hearing loss, hearing aid (HA) use, the length of the interval between implantations, hearing results before CI2 and the performance level with the first CI when counselling patients whether a second CI would be successful.^{9–19}

The majority of literature on factors affecting CI outcomes is about unilateral implantation. In 2009, Roditi et al²⁰ presented a prediction model for unilateral CI performance in postlingually deafened adults based on duration of any hearing loss in the CI ear, preoperative speech understanding in quiet and the length of severe to profound hearing loss in either ear. With their model, they could predict 60% of the variance in postoperative consonant-nucleus-consonant (CNC) scores. Our research group recently performed a systematic review to determine whether similar factors play a role in the success of sequential bilateral implantation as in unilateral implantation.²¹ We included ten papers on the effect of age, duration of hearing loss, time between implantations, preoperative hearing, aetiology of hearing loss, hearing aid use and duration of follow-up on sequential CI performance.^{4,9,10,12,13,17,22–25} Based on the best evidence available to date, advanced age, a long duration of deafness or a long interval between implantations does not necessarily lead to poorer sequential cochlear implantation outcome. The performance level with the first CI may be an important predictor for sequential implantation performance, but, to our knowledge, has only been examined in two studies.^{10,26} Unfortunately, the included studies were heterogeneous, had relatively low sample sizes, and the influence of a certain prognostic factor was often a secondary outcome of the study.²¹ It was therefore rather difficult to draw straightforward conclusions.²¹

The aim of this study was to contribute to filling this gap in the existing literature. We retrospectively utilised a database with a large number of sequentially implanted adult CI recipients to determine which preoperative factors are related to sequential cochlear implantation outcome. This led to the development of a prediction model based on the factors that were significantly correlated to auditory performance with a second CI. Knowing which factors are related to sequential cochlear implantation outcome will help CI

Keypoints

- Advanced age or a long interval between implantations does not necessarily lead to poor CI2 results.
- Patients who are successful HA users before CI2, who have a low PTA before CI2, a high CNC phoneme score with CI1 and a limited length of hearing loss before CI2, are likely to be successful CI2 recipients.

teams to more accurately counsel patients who are considering sequential implantation.

2 | METHODS

2.1 | Ethical consideration

The study was performed according to the principles expressed in the Declaration of Helsinki. The study was recognised as negligible risk and was granted an exemption from the Human Ethics Committee of the University of Western Australia (Reference number RA/4/1/8931).

2.2 | Study design and participants

This retrospective chart review was conducted within the CI audiology service managed by the Ear Science Institute Australia (ESIA). All patients who received a CI at the following affiliated hospitals: St John of God Hospital, Subiaco Private Hospital, Osborne Park Hospital and Sir Charles Gairdner Hospital in Perth were considered eligible for inclusion. The implant centre at ESIA is the largest in Western Australia, conducting approximately 12% of all cochlear implants in Australia, and its patients can be considered representative of an adult implant population. Comprehensive clinical data, including patient characteristics, implant details and pre- and post-surgical test results, have been collated since the start of the programme, and stored in a secure database. All postlingually deafened adult patients (≥ 18 years of age at the moment of the first implantation), who had undergone sequential cochlear implantations between 19 June 1990 (first cochlear implantation of the database) and 14 March 2016 were included in this study. The study outcome measure was the 12-month CNC phoneme score (speech intelligibility in quiet at 65 dB SPL). Patients were only excluded if this measure was

missing from the database and patient file. Two authors, YS and TH, verified whether the data in the database corresponded to the data in the patient records in the hospitals and adjusted the database if necessary. The data gathered were patients' gender, age of onset of any degree of hearing loss (age at which patients could remember their hearing loss started or when they started to use hearing aids), age at implantation of the first CI (CI1) and the second CI (CI2), side of first implantation, duration of deafness before CI1 and CI2 in each ear, interimplantation interval duration, origin of hearing loss for both ears, HA use, comorbidity expressed as the Charlson score (0 = no comorbidity, 24 = maximum comorbidity score)²⁷ and preoperative hearing details (pure tone average (0.5, 1, 2 kHz) in each individual ear and maximum speech intelligibility (CNC phoneme score) in each ear with and without HAs and with wearing CI1 only).

2.3 | Study outcome

The study outcome measure was the CNC phoneme score (%) with CI2. A full list of 25 words was presented in quiet at a fixed level of 65 dB SPL, from a speaker in front of the patient at 1 m distance. The outcome measure is the percentage phonemes repeated correctly. The test was performed 12 months after the second implantation.

2.4 | Data analysis

The statistical analyses were performed with IBM SPSS Statistics for Windows version 24.0 (IBM Corp. Armonk, NY: IBM Corp.). The data were, overall, normally distributed, and means, standard deviation and ranges are displayed in the tables. We used a multiple imputation technique to account for the missing values in our database. Only 2.8% of the data were missing, including all patient characteristics (Table 1) and hearing test outcomes. Ten imputations were used.²⁸ To analyse which variables were correlated to the study outcome, we performed univariate linear regression analyses. A correlation R is considered very weak when $R < 0.3$, weak when $R = 0.3$ -0.5, moderate when $R = 0.5$ -0.7, strong when $R = 0.7$ -0.85, very strong when $R = 0.85$ -0.95 and extremely strong when $R > 0.95$.^{29,30} Subsequently, we identified the variables that were significantly correlated to the outcome and entered these variables into a backward multiple linear regression analysis. This latter method analyses which factors are actual predictors for sequential cochlear implantation outcome and can be used to create a predictive model. The accuracy of the model is presented as the explained variance R^2 (<10% = very weak, 10%-25% = weak, 25%-50% = moderate, 50%-75% = strong, 75%-90% = very strong, >90% = extremely strong).^{29,30} We will present the accuracy of the model based on the imputed data and based on the original data.

3 | RESULTS

3.1 | Patients

A total of 142 adult patients received bilateral cochlear implants between 19 June 1990 and 16 March 2016. 50 patients did not

TABLE 1 Patient characteristics

	N	%	
Male	47	51.1	
Female	45	48.9	
Hearing aid use before CI1 in first implanted ear; yes (n = 91)	81	88.0	
Hearing aid use before CI2 in second implanted ear; yes (n = 92)	87	94.6	
Side of first implant			
Right	38	41.3	
Left	54	58.7	
Charlson score for comorbidity (n = 91) ¹³			
0	46	50.0	
1	16	17.4	
2	15	16.3	
3	5	5.4	
4	2	2.2	
5	4	4.3	
6	2	2.2	
7	1	1.1	
	Mean	SD	Range
Age at start hearing loss (y)(n = 90) ^a	29	19	0-66
Age at CI1 (y) (n = 92)	58	15	20-85
Age at CI2 (y) (n = 92)	61	15	21-87
Length of hearing loss before CI1 in this ear (y) (n = 90)	28	18	0.5-75
Length of hearing loss before CI2 in this ear (y) (n = 90)	32	19	0.7-80
Interval between implantations (y) (n = 92)	3.2	3.3	0.4-21
PTA 1st implanted ear preoperatively (dBHL) (n = 90)	104	17	52-120
PTA 2nd implanted ear preoperatively (dBHL) (n = 91)	90	17	48-120
CNC phoneme score before CI1 with 1st implanted ear (%) (n = 77)	17	18	0-72
CNC phoneme score 1 y post-CI1, with CI1 (%) (n = 86)	74	17	15-97
CNC phoneme score before CI2 with 2nd implanted ear (%) (n = 85)	23	23	0-72
CNC phoneme score 1 y post-CI2, with CI2 (%) (n = 92)	68	22	0-93

SD, Standard deviation; CNC, Consonant-nucleus-consonant; PTA, Pure tone average (0.5,1,2 kHz).

^aAge at which patients could remember their hearing loss started or when they started to use hearing aids.

meet the inclusion criteria; 34 patients had a prelingual deafness (significant hearing loss before the age of 3.5 years old), four patients had received their implants simultaneously during one surgery, and in 12 cases, the 1-year postoperative CNC phoneme score was incomplete. The remaining 92 patients were included. The patient characteristics are summarised in Table 1. There

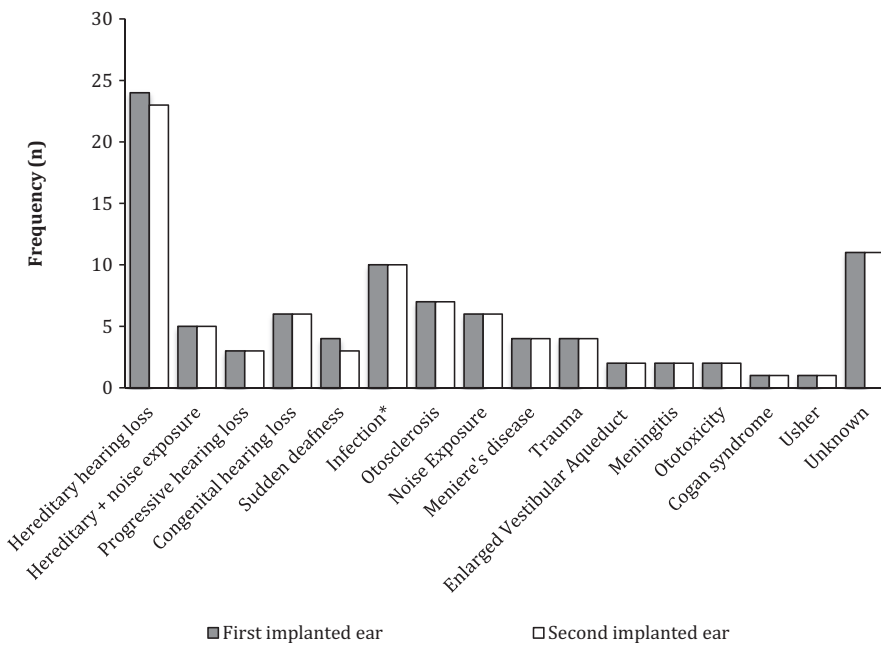


FIGURE 1 Origin of hearing loss.
*Infection encompasses, for example, measles, mumps, polio and rubella

TABLE 2 Correlations between preoperative variables and maximum consonant-nucleus-consonant (CNC) phoneme score in quiet with CI2

	R	Unstandardised B	P	95% confidence interval
Gender	0.02	0.92	0.84	−8.0-9.9
Hearing aid use before CI1 in this ear (yes)	0.15	10.7	0.14	−3.7-25.2
Hearing aid use before CI2 in this ear (yes)	0.46	44.1	0.00 ^a	26.4-61.8
Side of implantation	−0.19	−8.42	0.07	−17.5-0.6
Charlson score for comorbidity ¹³	−0.08	−1.0	0.47	−3.8-1.8
Age at start any hearing loss (y) ^a	0.14	0.16	0.20	−0.1-0.4
Age at start hearing loss in first implanted ear (y)	−0.04	−0.05	0.68	−0.3-0.2
Age at start hearing loss in second implanted ear (y)	0.13	0.14	0.22	−0.1-0.4
Length of hearing loss before CI1 (y)	−0.09	−0.11	0.40	−0.4-0.2
Length of hearing loss before CI2 (y)	−0.25	−0.29	0.02 ^a	−0.5-−0.1
Age at CI1 (y)	−0.19	−0.27	0.08	−0.6-0.0
Age at CI2 (y)	−0.17	−0.25	0.11	−0.6-0.1
Interval between implantations (y)	0.12	0.75	0.28	−0.6-2.1
Preoperative PTA before CI1 (dB HL)	−0.12	−0.15	0.26	−0.4-0.1
Preoperative PTA before CI2 (dB HL)	−0.27	−0.34	0.01 ^a	−0.6-−0.1
CNC phoneme score before CI1 with 1st implanted ear (%)	0.01	−0.01	0.92	−0.26-0.29
CNC phoneme score 1 year post-CI1, with CI1 (%)	0.21	0.29	0.05 ^a	−0.00-0.57
CNC phoneme score before CI2 with 2nd implanted ear (%)	0.18	0.17	0.11	−0.04-0.38

PTA, pure tone average (0.5, 1, 2 kHz).

^aAge at which patients could remember their hearing loss started or when they started to use hearing aids.

were approximately equal numbers of males and females. A vast majority of the patients used a HA before CI1 (88%) and even more before CI2 (95%). Most patients received their first CI on their left side (59%). Current clinical practice is for the worst hearing ear to be implanted first, which explains the difference in preoperative hearing results before CI1 and CI2. The length of hearing loss before CI2 is not the same as the length of hearing loss before CI1 plus the interval between implantations, because

the hearing loss may not have started at the same age in both ears.

The cause of hearing loss was extracted from all patient files. In many patients, a cause could be identified. However, when the cause was not clear, we described the progression of hearing loss, if known (eg, “sudden deafness,” or “progressive hearing loss”). We divided the origin of hearing loss into 16 categories (Figure 1). One patient had a different cause of hearing loss for each ear.

3.2 | Correlations

Table 2 shows the correlations between 18 preoperative factors and the study outcome. Only four factors correlated significantly with the postoperative CI2 CNC phoneme score. These factors were as follows: HA use before CI2 in this ear, length of hearing loss before CI2, preoperative pure tone average (PTA) before CI2 and the CNC phoneme score measured 12 months after CI1. We excluded “cause of hearing loss” from the analysis, because of the heterogeneity of this factor. Figure 2 shows the correlation between two predictive factors and CNC phoneme score for CI2.

3.3 | Predicting sequential CI outcome

Backward stepwise multiple regression analysis showed that three factors were significant contributors to predict the outcome of a sequential CI: Hearing aid use before CI2 in the second ear, the length of hearing loss before CI2 in the second ear and the CNC

phoneme score with CI1 at 65 dB SPL after 12 months of unilateral CI experience.

With this information, the following equation could be created:

Predicted CNC phoneme score with CI2 (%) = $16 + (44 * \text{HA use before CI2 (yes)}) - (0.22 * \text{length of hearing loss before CI2 (years)}) + (0.23 * \text{CNC phoneme score with CI1 (\%)})$.

We applied this model to the study population for internal validation. Figure 3 displays the predicted and the actual CNC phoneme scores with CI2. For the actual CNC phoneme score, the mean was $68\% \pm 22\%$ (SD). For the predicted CNC phoneme score, the mean was $68\% \pm 12\%$ (SD). The model based on the original data has a moderate accuracy of $R = 59\%$, $R^2 = 35\%$. The model based on the imputed data is $R = 55\%$, $R^2 = 30\%$.

The factor HA use appeared to play an important role; however, as it was based on only five patients, we repeated the analysis above after exclusion of this factor. Subsequently, the following equation could be created, this time including the factor preoperative PTA in CI2:

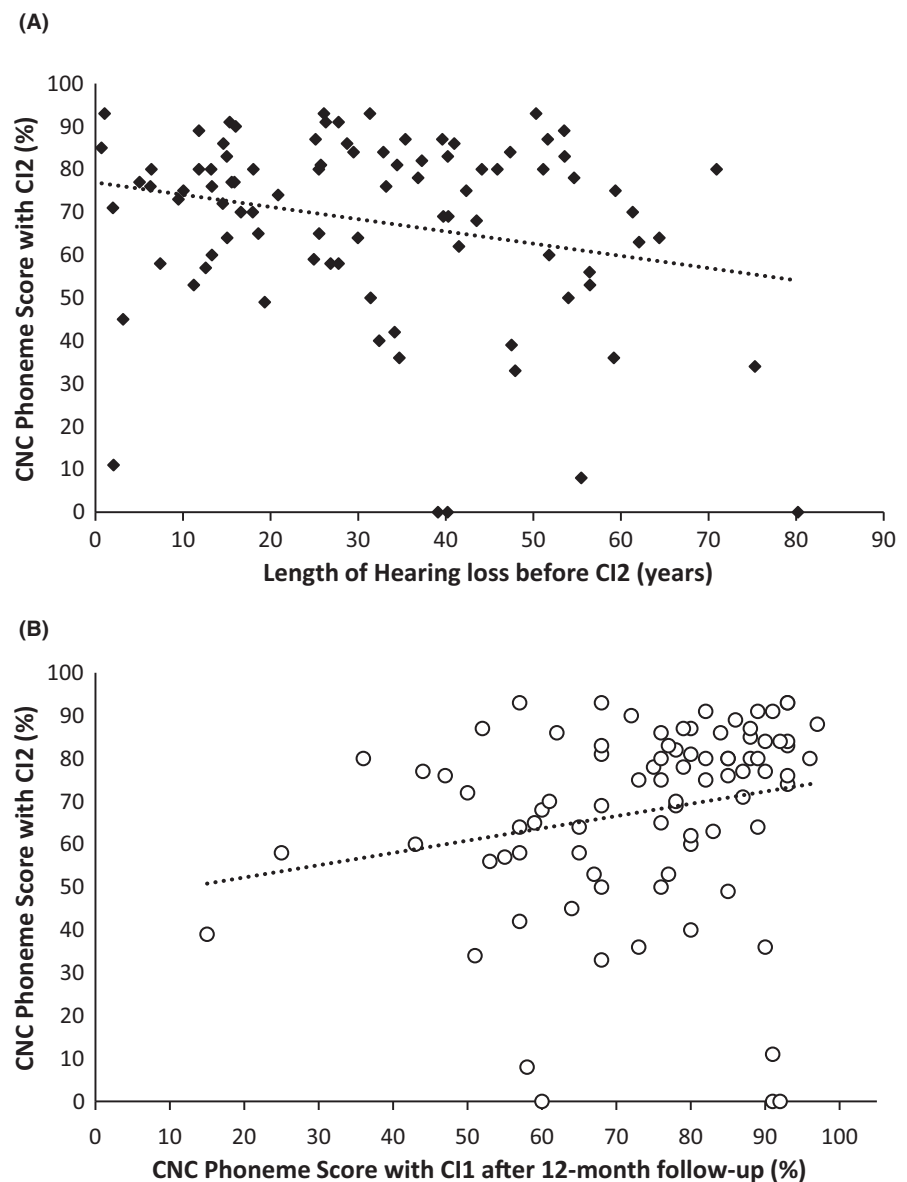


FIGURE 2 A, Length of hearing loss before CI2 vs consonant-nucleus-consonant (CNC) Phoneme Score with CI2 after 12-mo follow-up ($n = 90$). B, CNC Phoneme Score with CI1 after 12-mo follow-up vs CNC Phoneme Score with CI2 after 12-mo follow-up ($n = 86$)

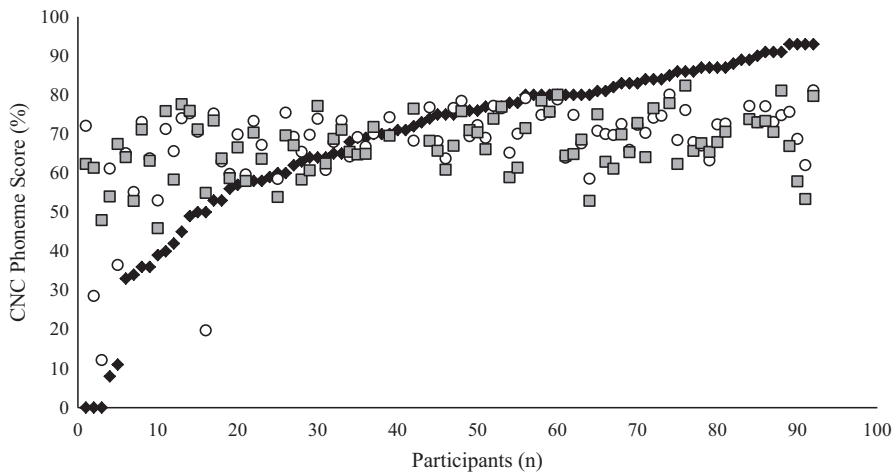


FIGURE 3 Predicted and measured consonant-nucleus-consonant (CNC) Phoneme score with CI2 at 65 dB SPL, 12 mo after CI2. ♦ CNC Phoneme Score with CI2 after 12 mo follow-up (n = 92); ○ Predicted CNC Phoneme Score with CI2 (equation 1, hearing aid factor included) (n = 84); ■ Predicted CNC Phoneme Score with CI2 (equation 2, hearing aid factor excluded) (n = 83)

Predicted CNC phoneme score with CI2 (%) = $82 - (0.17 * \text{length of hearing loss before CI2 (years)}) - (0.27 * \text{PTA in second implanted ear before CI2 (0.5, 1, 2 kHz)}) + (0.20 * \text{CNC phoneme score with CI1 (\%)})$.

For the predicted CNC phoneme score, the mean was $67\% \pm 8\%$ (SD). The model based on the original data has a weak accuracy of $R = 38\%$, $R^2 = 15\%$. The accuracy of the model based on the imputed data is $R = 35\%$, $R^2 = 12\%$.

4 | DISCUSSION

4.1 | Synopsis

The aim of this study was to determine which preoperative factors affect performance with a second CI after sequential cochlear implantation and to create a mathematical model to predict speech intelligibility in postlingually deafened adult patients undergoing sequential cochlear implantation. This model was based on patient characteristics (Table 1) identified through retrospective chart review of included patients.

One of the key factors that appeared to determine the success of sequential bilateral cochlear implantation was wearing a HA before CI2, although only five of the 92 patients did not wear a HA before CI2. All five patients did not benefit from a HA, due to the severity of the hearing loss. This finding can be explained as follows: when a patient is a successful HA recipient before CI2, it is likely that he/she will perform well with an implant in that ear. This does not imply that every candidate for a second implant should wear a hearing aid; in some cases, it will have no benefit. As this factor appeared to play such an important role based on the results of only a small portion of the group, we created a second model after exclusion of the factor HA use before CI2. However, the accuracy of the second model was considerably lower than that of the original model.

Prolonged duration of hearing loss before CI2 was a negative predictor and a high CNC phoneme score with CI1 a positive predictor for sequential cochlear implantation performance. When we removed the HA factor from the regression analysis, preoperative

PTA before CI2 also became an independent predictor for CI2 performance.

Our data also showed that several factors were not related to CI2 outcome, including patient's age, the length of the interval between implantations, the length of hearing loss before CI1 and a patient's comorbidity. This information is counterintuitive and is as valuable as knowing which factors are related to good or poor outcome.

4.2 | Strengths and weaknesses

A strength of this study is the high number of participants. A large study population is essential to perform a stepwise linear regression analysis and increases the internal validity of a study. Furthermore, the study has a low number of missing data and contains a large amount of information on each patient. We used a universally applied study outcome, which makes it possible to generalise our findings to other countries and studies. Literature has shown that bilateral cochlear implantation helps to restore sound localisation and improves hearing in noise.¹⁻⁸ Unfortunately, our patients did not undergo any specific binaural hearing tests. One may assume that better speech understanding in quiet in both ears will lead to better spatial hearing capabilities, but we could not prove this with the data available to us. Other weaknesses of the study are the retrospective design and that fact that the study was subject to selection bias. Patients may not have received a second CI in the past because the CI team had decided that a second CI would probably not be beneficial. It is most likely that the performance level with CI2 is not only affected by preoperative factors, but also by perioperative and post-operative aspects such as surgical technique (approach, traumatic insertion, use of protective/lubricant drops)^{31,32} and participation in postoperative auditory rehabilitation.³³⁻³⁵ The aim of this study was, however, to create a model based purely on preoperative characteristics. In general, the internal validity of a model could also be tested by splitting the database randomly and applying the model to the other half of the participants. However, with the amount of factors we analysed, the number of patients in the database was not

sufficient to perform such a test. External validity could be tested by applying the model to other databases. This would be interesting for further research.

4.3 | Comparison with the literature

A few other retrospective studies reviewed the influence of preoperative patient factors on sequential cochlear implantation outcome. In 2016, Boisvert et al.¹⁰ performed a study with 67 patients. They analysed the effect of six preoperative factors. As in our study, they found that the phoneme score with CI1 was an important, and in their study, the only significant predictor for performance with a second CI. In contrast to our findings, they did report a negative correlation between age and sequential cochlear implantation outcome, but all patients included were above the age of 50 years.

Other studies all had small sample sizes of 10-29 patients and reviewed a maximum of five different factors per study.^{4,9,12,13,23-25} There were several similar outcomes as in the current study. For example, age at implantation was not significantly correlated to sequential cochlear implantation outcome according to Zeitler et al and Boisvert et al.^{9,25} The degree of hearing loss PTA before implantation was not significantly correlated to sequential cochlear implantation outcome according to Boisvert et al.^{9,10} Furthermore, Reeder et al.¹³ reported that a prolonged duration of deafness before CI2 was a predictor for poor sequential cochlear implantation performance.

This analysis of 18 preoperative variables in a large study population is a contribution to the existing literature on expectations of sequential cochlear implantation performance. With the rising amount of adult patients being implanted bilaterally, the amount of data will keep growing and group results will become more able to provide accurate predictions. Our data showed that advanced age or a long interval between implantations does not necessarily lead to poor CI2 results. On the other hand, patients who are successful HA users before CI2, who have a low PTA before CI2, a high CNC phoneme score with CI1 and a limited length of hearing loss before CI2, are likely to be successful CI2 recipients. These findings may assist CI teams in providing evidence-based advice to their postlingually deafened adult patients who are interested in a second CI. For future research, it would be helpful if spatial hearing tests and localisation tests would become part of the routine follow-up in CI centres. Ultimately, the purpose of bilateral implantation is to restore binaural hearing and it would be interesting to study which preoperative factors eventually really affect binaural performance.

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CONFLICT OF INTEREST

The authors report no conflict of interests.

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